

Case Report

Noncommunicating Spinal Extradural Meningeal Cyst in Thoracolumbar Spine

Il Sup Kim, M.D., Jae Taek Hong, M.D., Byung Chul Son, M.D., Sang Won Lee, M.D.

Department of Neurosurgery, St. Vincent's Hospital, The Catholic University of Korea, Suwon, Korea

Spinal extradural meningeal cyst has been rarely reported, whose etiologies are assumed to be the communication of cerebrospinal fluid (CSF) between intradural subarachnoid space and cyst due to the congenital defect in dura mater. Although the CSF communication due to this defect can be found, in most case, few cases in which there is a lack of the communication have also been reported. We report a case of the huge extradural meningeal cyst occurring in the thoracolumbar spine (from T10 to L2) where there was a lack of the communication between the intradural subarachnoid space and cyst in a 46-year-old man who presented with symptoms that were indicative of progressive paraparesis and leg pain. The patient underwent laminectomy and cyst excision. On intraoperative findings, the dura was intact and there was a lack of the communication with intradural subarachnoid space. Immediately after the surgery, weakness and leg pain disappeared shortly.

KEY WORDS : Meningeal cyst · Noncommunicating · Cerebrospinal fluid · Thoracolumbar spine.

INTRODUCTION

Spinal extradural meningeal cyst (SEMC) has been reported very rarely, accounting for approximately 1-3% of all primary spinal space-occupying lesions^{6,13}. Nabors et al classified these cysts into three types : extradural meningeal cysts without spinal nerve root fibers (Type I); spinal extradural meningeal cysts with spinal nerve root fibers (Type II, Tarlov cyst); and spinal intradural meningeal cysts (Type III)¹⁶. Type I is further divided into extradural arachnoid cysts (Type IA) and sacral meningoceles (Type IB)¹⁶. In most of the reported cases of Type IA SEMC, there was a communication of the CSF with intradural subarachnoid space through dural defect^{1,3-5,7,10,11,16,20}. These dural defects are of congenital origin which are mostly associated with neural tube defects. But some acquired factors such as inflammation, trauma, surgery, or repeated lumbar puncture might cause dural defect^{6,8,15,18}. We preoperatively performed delayed myelo-computed tomography (CT), thin slice axial con-

structive interference in steady state magnetic resonance imaging (CISS-MRI), but could not observe the communication between the cyst and intradural subarachnoid space. To our knowledge, only one case of Type IA SEMC in which there was a lack of the communication with intradural subarachnoid space has been reported up to present. This is a very rare case. We experienced the improvement of symptoms using a cyst excision. Here, we report our case with a review of literature¹².

CASE REPORT

History and Examination

A 46-year-old man presented with a 10 years history of back pain, newly developed right leg pain, and gait disturbance. There was no history of trauma. Ten years ago, he suffered from spontaneously developed back pain and was treated with physical therapy. On admission, he could not walk by himself due to both leg weakness.

A physical examination did not reveal any notable findings except back pain and both leg weakness. The straight leg raising test was normal and deep tendon reflexes were increased. The motor powers of both leg foot dorsi-flexion, and foot plantar flexion were checked to be grade 4/5. Sensory, bladder, and bowel functions were normal. On electromyogram (EMG),

• Received : March 31, 2010 • Revised : May 9, 2010

• Accepted : December 31, 2010

• Address for reprints : Jae Taek Hong, M.D.

Department of Neurosurgery, St. Vincent's Hospital, The Catholic University of Korea, 93-6 Ji-dong, Paldal-gu, Suwon 442-723, Korea

Tel : +82-31-249-7190, Fax : +82-31-245-5208

E-mail : jatagi15@paran.com

there were findings suggestive of bilateral lower thoracic myelopathy. On somatosensory evoked potential (SEP), there were suggestive of central conduction delay above the thoracic area.

A thoraco-lumbar MRI revealed the presence of a large extradural cystic lesion with a multiple septations between T10 to L2 levels. To confirm the communication between the cystic lesion and intradural subarachnoid space, a CISS-MRI and a 6-hours delayed myelo-CT were performed. However, there was a lack of the communication between cystic lesion and intradural subarachnoid space (Fig. 1). Our case corresponded to type IA based on Nabors classification.

Operation

Total laminectomy from T10 to L1 were performed under intraoperative monitoring system. Under the surgical vision, the thinned lamina with an erosion could be confirmed. The laminectomy was performed and ligament flavum was removed. Then, the cyst with a translucent wall was observed (Fig. 2). Cyst was punctured and CSF-like clear fluid was aspirated. The cyst was easily dissected from outer surface of thecal sac and it was removed totally using a piecemeal. During cystectomy, there was a lack of the communications, such as dural defect, arachnoid pedicle, or fistula, between the cyst and dura mater. Following a complete removal of the cyst, Valsalva maneuver was performed. There was no evidence demonstrating CSF leaking. Once cyst decompression was done intraoperatively, the improvement of the amplitude of intraoperative SEP was observed compared with preoperative SEP amplitude.

Postoperative Course

On postoperative MRI scans, a total obliteration of the cyst was observed. A slight expansion of the spinal cord which was compressed due to the cyst was also observed (Fig. 3) at the time of postoperative 6 months follow up MRI. The histologic examination revealed the consistency of meningeal cyst which was characterized by fibrous connective tissue (Fig. 4). From immediately after the surgery, paresthesia and paraparesis of the right extremities were improved in the patient, and the patient was able to walk by himself. A surgical wound was well healed without CSF leaking. The patient was discharged without any notable events.

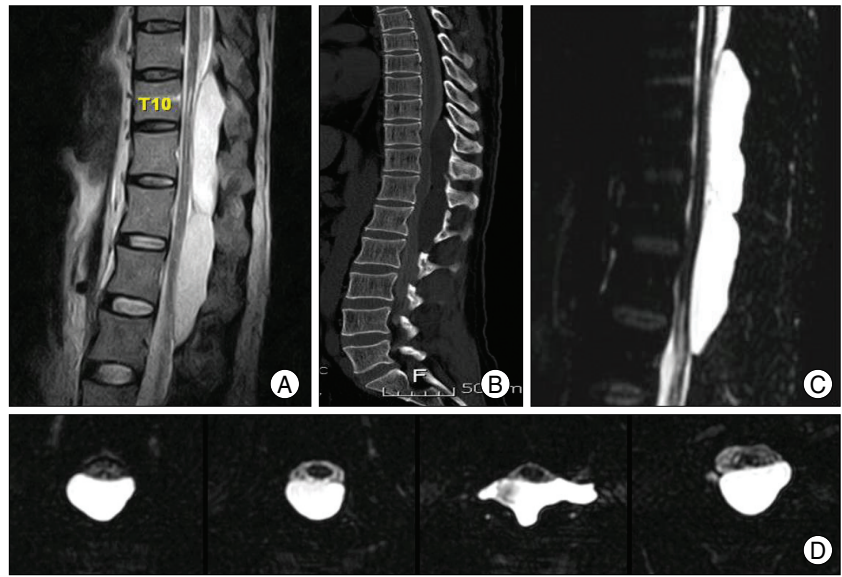


Fig. 1. Preoperative image findings. Sagittal T2-weighted magnetic resonance image (MRI) of thoracolumbar spine reveals a septated cystic lesion at the posterior extradural space at the T10-L2 level with high signal change in spinal cord (A). Six-hour delayed myelo computed tomography (CT) reveals no cerebrospinal fluid (CSF) communication between intradural subarachnoid space and cyst (B). Sagittal (C) and 3 mm thin slice axial (D) constructive interference in steady state magnetic resonance imaging (CISS-MRI) also reveal no cerebrospinal fluid (CSF) communication between intradural subarachnoid space and cyst.



Fig. 2. Intraoperative photograph shows a large extradural meningeal cyst arising from the dorsal aspect of the spinal canal. The cyst is being dissected from dorsal surface of thecal sac and there is a lack of the communications with intradural subarachnoid space.

DISCUSSION

Intradural cyst occurs most frequently in the middle to lower part of thoracic vertebrae (67%), and it also occurs in the lumbosacral area (20%), the thoracolumbar area (9%) and the cervical area (4%)^{4,14,19}. The lesions are mostly located dorsal to the intervertebral column and then extended to the intervertebral foramen. It shows a male predilection and symptoms usually occur during the 2nd decade^{3,19}. Most of the patients persistently present with rigid or flaccid paralysis of the extremities. A local back pain is also one of the common symptoms, but the sensory symptom was not clear^{3,19}. In some cases, the symptoms disappear for several



Fig. 3. Postoperative 6 months sagittal T2-weighted MRI reveals totally obliterated cyst with slightly expanded spinal cord.

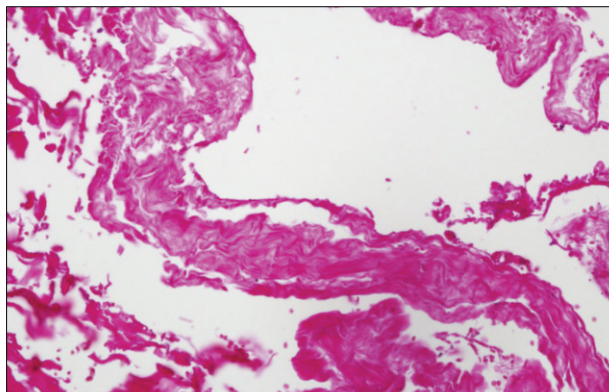


Fig. 4. The histologic examination reveals the consistency of meningeal cyst which is characterized by layered collagenous fibers and a membrane with flat lining cells. H & E, original magnification $\times 100$.

years. In other cases, the symptoms can be aggravated due to the posture or Valsalva maneuver^{3,14,19}.

On computed tomography (CT) scans and a simple X-ray, there can be characteristic imaging features such as the erosion of posterior lamina, pedicle and vertebral body, the extension of spinal canal, the extension of intervertebral foramen, the posterior scalloping of vertebral body and the extension of interpedicular space. A kyphoscoliosis can also occur^{3,4,7,20}. Through myelography or myelo-CT, the communication with subarachnoid space can also be confirmed^{7,11,19}. Kendall et al. and Nabors et al. showed in their series that only half of the extradural meningeal cysts initially exhibit opacification; on delayed studies nearly 100% will opacify^{9,16}. Using these methods, most cases of the CSF communication point between SEMC and intradural subarachnoid have been reported to be found. In some cases in

which an accurate localization of the CSF leaking point is difficult or those of noncommunicating or spontaneous closed cyst, as shown in the current case^{6,20}. To clearly identify CSF communication point, a preoperative work-up using CISS MR imaging technique has been reported to be helpful because of high spatial resolution and excellent contrast between CSF and solid structure¹⁷. Despite the use of these test regimens in this case, however, the definite communication of the CSF could not be found.

SEMC is assumed to occur because of spinal dysraphism or trauma, congenital diverticulum of the dura, or herniation of the arachnoid membrane through a congenital dural defect^{2,18}.

To explain the pathophysiology of cyst enlargement, the active fluid secretion theory and pulsatile CSF dynamic theory have been hypothesized. On histopathologic examination, they have been found to have no secretory functions. Therefore, it can be inferred that a pulsatile CSF dynamic theory is a more plausible theory^{14,19}. If pulsatile CSF is imported to the cyst via a ball valve mechanism and the pressure is lowered, the outlet is closed at the neck of a cyst^{14,19}. According to the Laplace law, the body of the cyst exerts a force on the neck sufficient to close the communication, because its radius and wall tension are greater. This mechanism then allows further enlargement, with persistent CSF pulsations¹². The current case is SEMC of noncommunication type, which has been reported very rarely. Some authors argued that osmosis caused cystic enlargement, but this theory has often been dismissed because most authors have indicated that the fluid content of cyst is the same as that of CSF^{3,8}. As mentioned above, the mechanism thought to responsible for the communication between the pre-exist-ing cyst and subarachnoid space to be enlarged based on Laplace law via a ball valve mechanism. Over the time, the communication would have been gradually closed and then disappeared. Also, there was myelopathy in spinal cord with pathologic spinal cord change in this case. We thought this myelopathy with myelomatic change would be secondary change of long standing spinal cord compression made by slow growing cyst.

In asymptomatic patients, the conservative treatment or observation rather than the surgical treatment is recommended¹⁸. Cases in which the neurologic deterioration or persistent pain due to SEMC are surgically indicated. Principles of the surgical treatment include a complete removal of the cyst, the obliteration of communication pedicle and the watertight repair of dural defect^{3,11}. According to the presence of adhesion or the location of lesions, however, a complete removal of the cyst may be impossible. There are many cases in which the lesions were extended to the intervertebral

foramen. In these cases, a massive amount of the hemorrhage can be concurrently present. In cases of large-sized cyst, the postoperative instability can be problematic. Even with a lack of a complete removal of the cyst, cases in which the treatment outcome was good have also been reported^{11,12,19}. The most problematic complication following SEMC surgery includes the recurrence of cyst. To prevent this, the communication with subarachnoid space should be identified and its dural defect should be sutured. A simple drainage of the cyst contents might temporarily alleviate the symptoms. As reported in the current case, SEMC of noncommunicating type can be managed by a single use of the cyst excision without a closure of the dural defect.

CONCLUSION

We have described a rare case of back, leg pain and leg weakness in patient with huge noncommunication SEMC in thoracolumbar region. In our case, there was a lack of the communicating dural defect on delayed myelo-CT, CISS MR image technique and under the surgical vision. This led to a tentative diagnosis of SEMC of noncommunicating type. With a surgical removal of the cyst, there was an improvement of the symptoms.

References

1. Bergland RM : Congenital intraspinal extradural cyst. Report of three cases in one family. *J Neurosurg* 28 : 495-499, 1968
2. Chang IC, Chou MC, Bell WR, Lin ZI : Spinal cord compression caused by extradural arachnoid cysts. Clinical examples and review. *Pediatr Neurosurg* 40 : 70-74, 2004
3. Cloward RB : Congenital spinal extradural cysts : case report with review of literature. *Ann Surg* 168 : 851-864, 1968
4. Congia S, Coraddu M, Tronci S, Nurchi G, Fiaschi A : Myelographic and MRI appearances of a thoracic spinal extradural arachnoid cyst of the spine with extra- and intraspinal extension. *Neuroradiology* 34 : 444-446, 1992
5. Dickson RA, Arabi K, Goodfellow J : Congenital spinal extradural cyst (lateral meningocele) simulating acute transverse myelitis. Report of a case. *J Bone Joint Surg Br* 60 : 412-415, 1978
6. Di Lorenzo N, Fortuna A, Guidetti B : Craniovertebral junction malformations. Clinicoradiological findings, long-term results, and surgical indications in 63 cases. *J Neurosurg* 57 : 603-608, 1982
7. DiSclafani A II, Canale DJ : Communicating spinal arachnoid cysts: diagnosis by delayed metrizamide computed tomography. *Surg Neurol* 23 : 428-430, 1985
8. Gortvai P : Extradural cysts of the spinal canal. *J Neurol Neurosurg Psychiatry* 26 : 223-230, 1963
9. Kendall BE, Valentine AR, Keis B : Spinal arachnoid cysts : clinical and radiological correlation with prognosis. *Neuroradiology* 22 : 225-234, 1982
10. Lee MC, Lee JK, Wee SC, Kim TS, Kim JH, Kim SH, et al. : Spinal extradural meningeal cyst - case report. *J Korean Neurosurg Soc* 27 : 407-411, 1998
11. Liu JK, Cole CD, Kan P, Schmidt MH : Spinal extradural arachnoid cysts; clinical, radiological, and surgical features. *Neurosurg Focus* 22 : E6, 2007
12. Liu JK, Cole CD, Sherr GT, Kestle JRW, Walker ML : Noncommunicating spinal extradural arachnoid cyst causing spinal cord compression in a child - case report. *J Neurosurg* 103 : 266-269, 2005
13. Lombardi V : Post-operative subdural hematoma with associated extradural arachnoid cyst of spine. Case report. *Acta Neurol (Napoli)* 25 : 123-125, 1970
14. McCrum C, Williams B : Spinal extradural arachnoid pouches. Report of two cases. *J Neurosurg* 57 : 849-852, 1982
15. Mummaneni PV, Pitts LH, McCormack BM, Corroo JM, Weinstein PR : Microsurgical treatment of symptomatic sacral Tarlov cysts. *Neurosurgery* 47 : 74-79, 2000
16. Nabors MW, Pait TG, Byrd EB, Karim NO, Davis DO, Kobrine AI, et al. : Updated assessment and current classification of spinal meningeal cysts. *J Neurosurg* 68 : 366-377, 1988
17. Nakagawa A, Kusaka Y, Jokura H, Shirane R, Tominaga T : Usefulness of constructive interference in steady state (CISS) imaging for the diagnosis and treatment of a large extradural spinal arachnoid cyst. *Minim Invasive Neurosurg* 47 : 369-372, 2004
18. Rabb CH, McComb JG, Raffel C, Kennedy JG : Spinal arachnoid cysts in the pediatric age group : an association with neural tube defects. *J Neurosurg* 77 : 369-372, 1992
19. Rohrer DC, Burchiel KJ, Gruber DP : Intraspinal extradural meningeal cyst demonstrating ball-valve mechanism of formation. Case report. *J Neurosurg* 78 : 122-125, 1993
20. Sato K, Nagata K, Sugita Y : Spinal extradural meningeal cyst: correct radiological and histopathological diagnosis. *Neurosurg Focus* 13 : ECP1, 2002